Bilateral Frontal Sinus Pericranii with an Intratabular Course

A Case Report

B. SCHENK, P.A. BROUWER

Radiology Department, Leiden University Medical Center; Leiden, The Netherlands

Key words: sinus pericranii, dural sinus, venous anomaly

Summary

Sinus pericranii is a rare venous anomaly, representing a transosseous connection between the intracranial venous system and the epicranial venous system. We present an unusual case of bilateral frontal sinus pericranii in a 12-year-old boy, with associated lacrimation. Instead of the usual short bridging vein between the intra- and extracranial venous circulation, in our case the veins connecting the superior sagittal sinus and the left superior ophthalmic/orbital vein coursed intratabularly over a distance of several centimeters. To our knowledge, such a course has not previously been reported in literature.

Introduction

Sinus pericranii is a rare venous anomaly, representing a transosseous connection between the intracranial venous system (e.g. dural sinuses) and the epicranial venous system. The condition was first described by Stromeyer in 1850 ¹. The classic presentation of a sinus pericranii is that of a child with a nonpulsatile soft tissue mass, that can be 'pressed away' and increases in size in situations of increased venous pressure, revealing its venous origin ^{1,2}.

The majority of sinus pericranii are located in the midline of the frontal region where they connect to the parasagittal sinus. However, other locations have also been reported ^{3,4}. Although most sinus pericranii occur as solitary lesions, up to 20% of patients are reported to have associated venous malformations elsewhere ^{5,6}. The exact mechanism by which sinus pericranii arise is unknown. The most widely

accepted theory is that of a developmental venous anomaly, possibly resulting from transient venous hypertension during the late phase of embryologic development ⁷. There have also been reports of acquired sinus pericranii attributed to direct head trauma ⁸. Most sinus pericranii are asymptomatic and the majority of patients seeking medical aid do this for cosmetic purposes. Only a small fraction of patients tend to experience discomfort or pain ^{7,9}. Although spontaneous thrombosis has been reported in some cases ¹⁰, the prognosis of untreated sinus pericranii is usually benign ^{7,9}. Lesions usually stay stable in size after puberty and have even been reported to regress spontaneously ¹¹.

Diagnosis and Treatment

The diagnosis of sinus pericranii can usually be suspected on clinical examination. The role of imaging is to confirm the clinical diagnosis, to rule out more serious pathology and to assess possible therapeutic options if desired ^{2,7,9} CTA and MRA are usually the first imaging modalities used as they can assess both the bony defect and the vascular anatomy of the lesion in a way superior to ultrasound or conventional radiography ^{10,12,13}. However, these imaging modalities do not provide information on the flow-dynamics of the lesion under investigation. This additional information is indispensable to differentiate a benign sinus pericranii from more serious pathology, such as, for instance, dural arteriovenous fistula 7. CTA and MRA certainly have an important role in ruling out the presence of a sinus pericranii when they are unremarkable. However, when abnor-

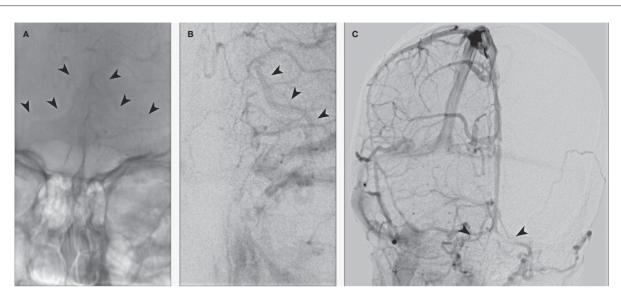


Figure 1 DSA+DA images, LICA+RICA injection. On both sides, the intratabular intra-osseous channels are clearly visible. Also, venous drainage through the sinus pericranii on both sides is depicted.



Figure 2 3d reconstruction of the superficial venous drainage through the angular-and facial veins on both sides. The vein on the right is slightly dilated. The vein on the left shows marked dilatation, consistent with the more prominent sinus pericranii on the left.

malities are present, in our opinion, additional DSA (or, in the future, 4dCTA) should be performed to assess flow dynamics. This is especially important if treatment is contemplated, when special care should be given to pattern of venous drainage ⁷. When only a small amount of blood from a relatively less important brain region is drained through the sinus pericranii, it can be safe to treat it either surgically or by endovascular intervention (coiling or embolization) ⁹. In cases where a significant amount of intracranial venous blood is drained through

the sinus pericranii, or when the source of this blood is a crucial brain area, the risk of either severe bleeding or brain ischemia through venous hypertension pose strong contraindications for treatment ⁷⁻⁹.

Case Report

We describe a case of an unusual type of sinus pericranii which, to our knowledge, has not previously been reported in literature. The pa-

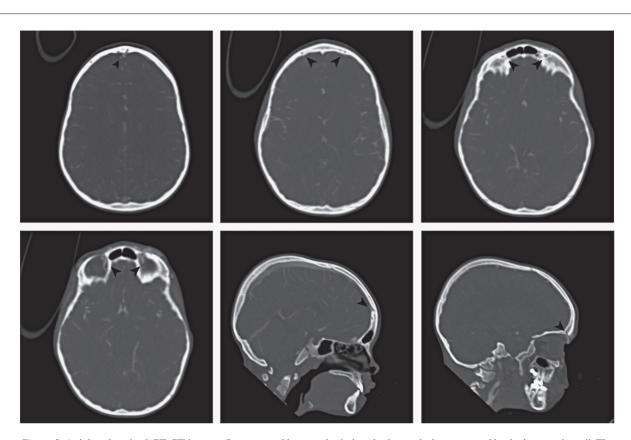


Figure 3 Axial and sagittal CE CT images. Sequence of images depicting the intratabular course of both sinus pericranii. The entry-point through the tabula interna is clearly seen, as well as the exit through the orbital roof into the orbit.

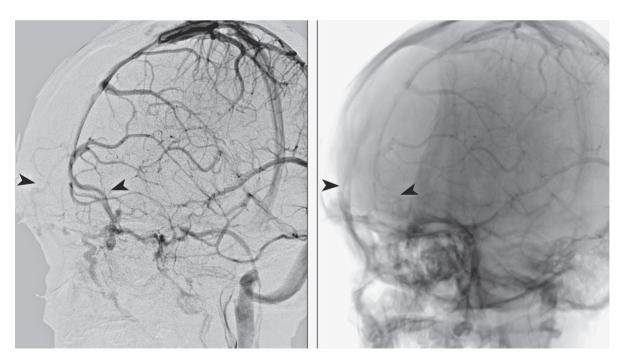


Figure 4 DSA + DA image, LICA injection. Full view of the venous drainage from the left side, depicting the connection of both sinus pericranii to the superior sagittal sinus, the intratabular course and the connection to the angular and facial veins on both sides.

tient is a 12-year-old boy, who presented with a soft bluish swelling located medially in the left frontal region. When venous pressure was raised (Valsalva maneuver, coughing, bending over) the swelling increased, with associated lacrimation from the left eye.

CTA and MRA performed in another hospital were initially reported as unremarkable. The patient was then referred to an ophthalmologist in our institution for analysis of the lacrimation disorder. Review of the previously performed CTA and MRA by our department revealed aberrant veins originating from the superior sagittal sinus and crossing the inner tabula on both sides through small bony defects. After coursing between the inner- and outer tabulae for several centimeters, the aberrant veins crossed the roof of the orbit to fuse with the left- and right superior orbital (or ophthalmic) veins, draining into the angular veins and subsequently outward. The aberrant vein on the left was markedly more pronounced then than the aberrant vein on the right. Additional DSA, performed to assess venous flow dynamics, showed the direction of venous blood flow to be from intracranial to extracranial. In this case, the sinus pericranii turned out to be part of an aberrant drainage path of the superior sagittal sinus through the left and right facial veins instead of the usual drainage path through the sigmoid sinus. After establishing the benign nature of the lesion, no further treatment was instigated.

Discussion

The case presented in this report is unusual in several ways.

First, instead of the usual short bridging vein between the intra- and extracranial venous circulation, in our case the veins connecting the superior sagittal sinus and the left- and right superior ophthalmic/orbital veins coursed intratabularly over a distance of several centimeters. To our knowledge, such a course has not previously been reported in literature.

The second unusual aspect is the clinical presentation, with excessive lacrimation on increased venous pressure. In this case, this phenomenon might be explained by the increased venous blood flow in the region of the left lacrimal gland.

Although some authors state that CTA or MRA alone are sufficient in confirming the clinical diagnosis of sinus pericranii ^{12,13}, it is our strong opinion that additional DSA (or, in the future, 4dCTA) provides invaluable information regarding the flow dynamics of the lesion under investigation and should therefore always be performed

Without DSA, it is virtually impossible to differentiate a benign sinus pericranii from potentially dangerous lesions such as dural arteriovenous fistulas unless a pulsatile arterial flow is observed in the lesion, especially with a long intra-osseous course of the sinus.

References

- 1 Stromeyer. About Sinus Pericranii (translation of original 850 text). Surg Neurol. 1993; 40: 3-4.
- 2 Lasjaunias PL, Berenstein A, Ter Brugge K. Surgical neuroangiography: Vol. 1: Clinical Vascular Anatomy and variations. 2nd ed. Berlin Heidelberg New York: Springer, 2001.
- 3 Koshu K, Takahashi S. Laterally situated sinus pericranii. Report of two cases with marked diploic veins. Neuroradiology. 1981; 21: 219-221.
- 4 Nishio A, Sakaguchi M, Murata K, et al. Lateral situated sinus pericranii. Case report. Surg Neurol. 1989; 32: 382-386.
- 5 Desai K, Bhayani R, Goel A, et al. Sinus pericranii in the frontal region: A case report. Neurol India. 2001; 49: 305-307.
- 6 Lasjaunias PL, Berenstein A, Ter Brugge K. Surgical Neuroangiography: Vol. 3: Clinical and Interventional Aspects in Children. 2nd ed. Berlin Heidelberg New York: Springer, 2007.
- 7 Gandolfo C, Krings T, Alvarez H, et al. Sinus pericranii: diagnostic and therapeutic considerations in 15 patients. Neuroradiology. 2007; 49: 505-514.
- 8 Kaido T, Kim YK, Ueda K. Diagnostic and therapeutic considerations for sinus pericranii. J Clin Neurosci. 2006; 13: 788-792.

- 9 Burrows PE, Konez O, Bisdorff A. Venous variations of the brain and cranial vault. Neuroimag Clin N Am. 2003; 13: 3-26.
- 10 Carpenter JS, Rosen CL, Bailes JE, et al. Sinus pericranii: clinical and imaging findings in two cases of spontaneous partial thrombosis. Am J Neuroradiol. 2004; 25: 121-125.
- 11 Rozen WM, Joseph S, Lo PA. Spontaneous involution of two sinus pericranii a unique case and review of the literature. J Clin Neurosci. 2008; 15: 833-835.
- Bigot JL, et al. Sinus pericranii: advantages of MR imaging. Pediatr Radiol. 2000; 30: 710-712.
 M, Fujimoto Y, Ikeda H, et al. Sinus pericranii: neuro-
- 13 M, Fujimoto Y, Ikeda H, et al. Sinus pericranii: neuroradiologic findings and clinical management. Higuchi Pediatr Neurosurg. 1997; 27: 325-328.

Barry Schenk, M.D. Fellow Interventional Neuroradiology Department of Radiology Leiden University Medical Center Albinusdreef 2 Leiden, 2333 ZA, Netherlands E-mail: b.schenk@lumc.nl